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Case Report

A case of spontaneous pneumomediastinum with unknown etiology

Timuçin Alar[1], Ahmet Sami Bayram[2], İsmail Ertuğrul Gedik[1]
[1]Thoracic Surgery Department, Çanakkale Onsekiz Mart University Medical Faculty, Çanakkale,
[2]Thoracic Surgery Department, Uludağ University Medical Faculty, Bursa, Turkey

Abstract

First described by Hamman in 1939, spontaneous pneumomediastinum (SPM) is a disease with the collection of air in the mediastinum without any underlying cause. It is usually seen in young males and its incidence is reported between 1/15000-1/25000 in different case series. We would like to present the case of a 16-year-old male patient who admitted to our hospital with the complaints of pleuritic chest pain and tenderness in the neck and was diagnosed as spontaneous pneumomediastinum. Patient spontaneously recovered without the need of an invasive procedure.

Keywords

Spontaneous Pneumomediastinum; Thoracic Surgery; Hamman's Sign

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Corresponding Author: Timuçin Alar, Thoracic Surgery Department, Çanakkale Onsekiz Mart University Medical Faculty, Çanakkale, Turkey.
E-Mail: timalar@yahoo.com
ORCID ID: 0000-0002-4719-002X

Introduction

Spontaneous pneumomediastinum (SPM) is a disease which was described for the first time by Hamman in 1939 and expressed as collection of air in the mediastinum without any underlying cause [1]. Usually, this disease is seen in young males and its incidence is calculated to be between 1/15000-1/25000 in different case series [2,3]. We aim to present the case of a 16-year-old male patient who admitted to our hospital with the complaints of pleuritic chest pain and tenderness in the neck and was diagnosed as spontaneous pneumomediastinum.

Case Report

A 16-year-old-male patient admitted to the outpatient clinics of the cardiology department of our hospital with the complaints of chest pain and tenderness in the neck that started approximately 24 hours before his admission. The physical examination and the posterior-anterior (PA) chest x-ray revealed Hamman's sign (Figure 1). He was referred to our outpatient clinics after a contrast-enhanced thoracic computerized tomography (CT) was requested. After 3 days from his first admission the patient was admitted for CT and then admitted to our outpatient clinics. He had a history of a chest pain with an onset 3 days before his admission and lasted for one day. The pain was pleuritic and localized retrosternally. He also had tenderness in the neck region which started simultaneously with the chest pain. He reported no fever or malaise. He did not have a history of respiratory tract infection, asthma, vomiting, cough with shear intensity, trauma, constipation or use of narcotic agents. His physical examination revealed that his blood pressure was 122/83 mm Hg, heart rate was 87/minute, body temperature was 36,3º C. There was no tenderness or crepitation in the cervical region. There weren't any abnormalities in the lung auscultation. In addition to the Hamman's sign found on the PA chest x-ray which was taken 3 days ago, the contrast-enhanced thoracic CT images revealed that there was mediastinal emphysema starting from the superior mediastinum and extending inferiorly (Figure 2). There was no finding at the CT suggesting mediastinitis and

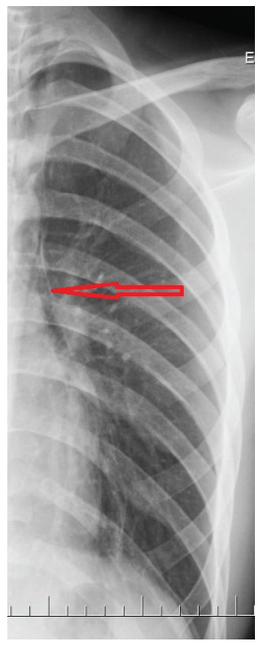

Figure 1.

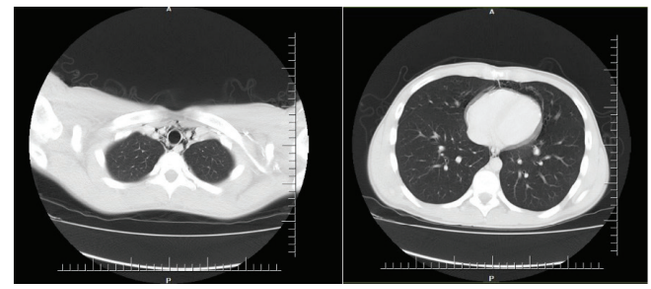

Figure 2.

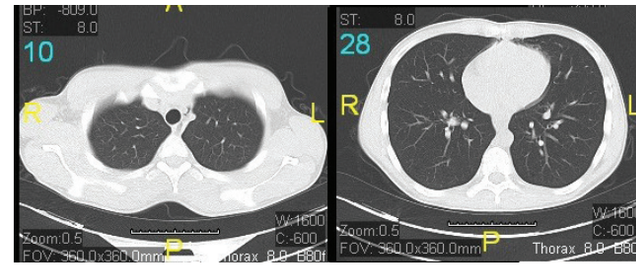

Figure 3.

the patient was diagnosed as SPM. In the blood analysis white blood cells were 7800/mm3 and the erythrocyte sedimentation rate was 8mm/hour. He was discharged and advised to be re-examined 3 days later. On his second examination 3 days later he had a history of vomiting the day before the second examination. He had no additional complaints and the physical examination revealed no abnormal findings but the patient was hospitalized in order to rule out a possible esophageal perforation. Oral ingestion was prohibited, fluid and electrolytes was replenished intravenously (IV). Cefazoline 1gr flacon 3x1 IV was administered as a prophylaxis for mediastinitis. Due to the possibility of the development of mediastinitis secondary to a possible esophageal perforation it was decided that performing an oral contrast-enhanced thoracic CT and follow up in the intensive care unit might be necessary. In order to establish these conditions patient was transferred to the Thoracic surgery clinics of the Uludağ University School of Medicine. Oral contrast- enhanced thoracic CT was performed and revealed no abnormalities. He didn't have fever or leukocytosis and ESR did not increase. A second oral contrast-enhanced thoracic CT was performed at his fourth day in thoracic surgery clinics and revealed that the mediastinal emphysema deteriorated. Thus, we did not find any suggestive findings for esophageal perforation or mediastinitis and oral ingestion of fluids was allowed. The patient has tolerated the oral food ingestion and was discharged from the hospital at the fourth day of admission and was invited for an outpatient clinic control examination 3 weeks afterwards. A final contrast-enhanced thoracic CT was performed at this outpatient clinics control examination. This CT revealed a complete resolution of the emphysema (Figure 3). We could not identify an etiological factor for the SPM throughout this whole medical work-up.

Discussion

SPM is seen more frequently in children and adolescents than in adults. The incidence of SPM is found to be between 1/14000-3/1000 in children despite differences in terms of age groups and comorbidities. It is thought that SPM incidence in children

is higher because of higher frequencies of asthma and respiratory tract infections which lead to severe coughing episodes [4]. Our patient was an adolescent which may be a supportive fact for the SPM diagnosis but he didn't have asthma or any other illness which may lead to prolonged coughing episodes.

It was postulated that several factors may have a key role in the development of SPM. Asthma, strenuous sports activities, diabetic ketoacidosis, labor, severe coughing or vomiting and the use of narcotic agents are considered to be the etiologic factors of SPM [5]. Barotrauma can be considered as the main factor in the pathophysiology of SPM. Barotrauma leads to increased intra-alveolar pressure and alveolar ruptures occur as a result in this increase in pressure. It is also considered that if these ruptures occur in close proximity with a pulmonary vessel or bronchi, air may travel through the peribronchovascular sheath and reach the mediastinum [4]. There was no evidence of barotrauma in our case.

History and physical examination are without a doubt the most important steps in the diagnosis of SPM. The typical symptom of SPM is the pleuritic chest pain which starts in the retrosternal area and distributes to the cervical region but it may also be asymptomatic. Dyspnea, dysphagia, dysphonia, change in the tone of the voice may also accompany the chest pain. The physical examination findings are crepitation at the cervical region that occurs due to the subcutaneous emphysema, cervical tenderness, decrease or absence of the respiratory sounds if SPM is accompanied with pneumothorax and crepitation sound which occurs synchronously with the heartbeat which is also known as Hamman's sign [6]. The definitive diagnosis of the SPM can be established with radiological findings. Hamman's sign which is the radiolucent silhouette of the heart seen on PA chest x-ray and/or mediastinal emphysema seen on the thoracic CT should be observed in order to achieve the diagnosis of SPM. It has been postulated that the PA chest x-ray is found to be normal in 30% of the patients with SPM and the thoracic CT is diagnostic in almost 100% of the cases [7]. Similar to the medical literature, our patient had a chief complaint of pleuritic chest pain which started retrosternally and distributed to the cervical region. He also had Hamman's sign on chest x-ray and mediastinal emphysema on the thoracic CT and thus he was diagnosed as SPM.

The patient was re-interviewed to reveal any possible cause of SPM. The patient was asked if he had had asthma or diabetes which was revealed to be absent. We have also evaluated the patient for strenuous sports activities and narcotic abuse since he was an adolescent which was revealed to be negative. He also mentioned that he did not have coughing symptom. The only positive symptom he had was vomiting.

The treatment of SPM is focused on treating the underlying disease and symptomatic relief. It should also be kept in mind that the entry of non-sterile air into the mediastinum might cause mediastinitis. It is suggested that oxygen inhalation therapy, administration of analgesics should be given and if the markers of inflammation increase in blood, antibiotherapy should also be administered to the patient [6]. We also used a similar treatment protocol in our clinics. But we do not agree to the suggestion of starting antibiotic prophylaxis for mediastinitis after an increase in the inflammatory markers in blood occurs; because of the mortality rate of the mediastinitis is still as high as 50% despite modern treatment. Because of that, we thought it would be more suitable for administrating prophylactic antibiotics as soon as possible. We also prohibited oral ingestion because of the possibility of an esophageal perforation.

SPM usually has a good prognosis. But several cases were reported in which the prognoses worsened quickly and some of these patients even required surgical intervention [8]. Our patient healed completely with conservative treatment.

In conclusion SPM is a disease which can be diagnosed easily without delay and can usually be treated conservatively. But SPM shares similar symptoms with other diseases which are seen more frequently than SPM. We believe that thoracic CT should be performed in order to achieve prompt and accurate diagnosis in any patient if there is a clinical suspicion of an underlying SPM because of the possibility of its complications that may easily cause mortality.

Scientific Responsibility Statement

The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

Animal and human rights statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No animal or human studies were carried out by the authors for this article.

Conflict of interest

None of the authors received any type of financial support that could be considered potential conflict of interest regarding the manuscript or its submission.

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